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Case Report

A Case of Direct Carotid-Cavernous Fistulae Successfully Treated by Bidirectional Double Catheter Technique: A Technical Note

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Objective: Ruptured carotid-cavernous aneurysms (CCAs) are known to result in direct carotid-cavernous fistula (CCF). Although endovascular treatment is recognized as the first-line treatment for direct CCF, obliteration is sometimes difficult because of the high-flow shunt. In this report, we present a case of a direct CCF treated by the combination of transarterial and transvenous approaches.

Case Presentation: A 57-year-old woman presented with conjunctival chemosis, exophthalmos, and tinnitus. Ophthalmological examination revealed increased intraocular pressure. DSA demonstrated a direct CCF due to a right ruptured CCA with retrograde shunted flow through the superior ophthalmic vein (SOV), superficial middle cerebral vein, basal vein of Rosenthal, and middle temporal vein. Two microcatheters were guided into the shunt segment from the internal carotid artery and SOV. In addition, a balloon catheter was placed at the neck of the aneurysm to assist coiling. Coil embolization for the CCF was performed using two microcatheters in the opposite direction, which enabled compact and tight packing of the shunt segment with only six coils. The CCF was eliminated. Two-year-follow-up MRA revealed no recurrence.

Conclusion: The bidirectional double catheter technique is a useful approach to obliterate a shunt in a short segment with minimal coils.

Keywords ▶ direct carotid-cavernous fistula, transvenous embolization, transarterial embolization

Introduction

Direct carotid-cavernous fistula (CCF) is a disease in which an abnormal shunt occurs between the internal carotid artery (ICA) and the cavernous sinus. This disease suddenly causes ophthalmoplegia, conjunctival chemosis, and exophthalmos. Although traumatic CCF is popular in direct CCF, some occurs through ruptured carotid-cavernous aneurysms (CCAs).[1)] Most of CCAs are asymptomatic; however, large or giant CCAs can result in cranial nerve palsy related to mass effects. Although the incidence of CCA rupture is reported low, ruptured CCAs induce direct CCF or rarely lead to subarachnoid hemorrhage.[2)] As treatments for direct CCF, embolization of aneurysm with detachable balloon or coils, flow diverter, and parent artery occlusion have been reported. However, shunt occlusion is sometimes difficult because of high-flow shunt.[2,3)] In this study, we report a case of direct CCF due to CCA rupture treated by coil embolization with the double-catheter technique, in which microcatheters were guided from two directions, the ICA and the superior ophthalmic vein (SOV), and led to complete shunt occlusion.

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Case Presentation

Patient: A 57-year-old woman.
Medical history: No history of head trauma.

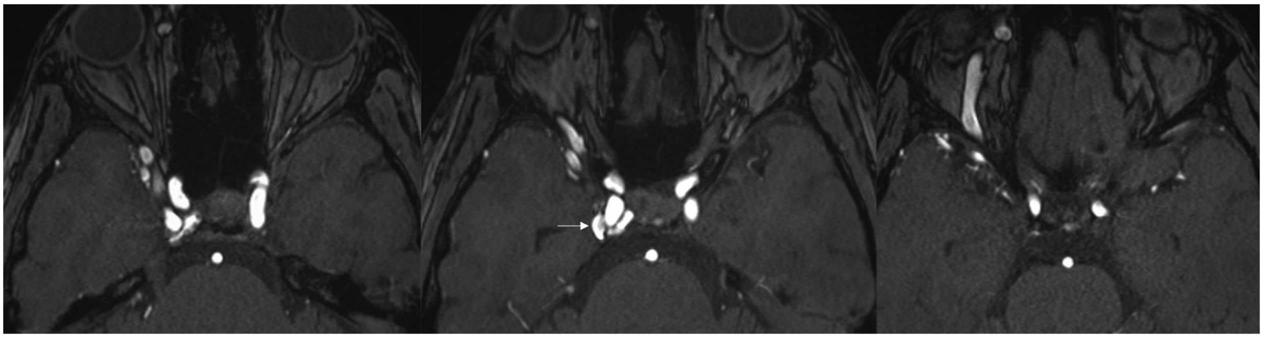

Fig. 1 MRI on admission. Time-of-flight-MRA on admission shows enlargement of the SOV and significant flow signal at the right cavernous sinus. The CCA is indicated by an arrow. CCA: carotid-cavernous aneurysm; SOV: superior ophthalmic vein

Present illness: She had a 1-year history of pulsatile right tinnitus. Right conjunctival chemosis and exophthalmos persisted for 3 months. She was referred to our department suspected of CCF with MRI and MRA at a local hospital.

Physical examination on admission: Consciousness was clear. Pulsatile right tinnitus, right conjunctival chemosis, and exophthalmos (right: 17 mm and left: 10 mm) were noted. Neither disturbance of eye movement nor diplopia was observed. Ocular pressure: right, 23 mmHg and left, 17 mmHg.

Radiological findings: MRI revealed no intracranial hemorrhage. Time-of-flight-MRA demonstrated a high-signal-intensity area in the cavernous sinus, enlarged right SOV, and a cavernous sinus aneurysm located in an area lateral to the ICA (**Fig. 1**). A high-flow shunt at the CCA was found on cerebral angiography (**Fig. 2A**–**2E**). Shunted flow drained to the facial and external jugular veins via the angular vein from the SOV, and a stenotic lesion of the angular vein was observed (**Fig. 2B** and **2D**, arrows). In addition, intracranial venous reflux to the superficial middle cerebral vein (SMCV) and basal vein of Rosenthal, and a venous drainage to the middle temporal vein were noted (**Fig. 2C** and **2D**). The intracranial cortical veins exhibited a pseudophlebitic pattern (**Fig. 2D**).

Endovascular treatment: The CCF was symptomatic with cortical venous reflux, and an increase in the ocular pressure for the right eye was observed. Therefore, early therapeutic intervention was conducted considering the risks of intracranial hemorrhage and loss of vision. Under general anesthesia, coil embolization of CCA through transarterial and transvenous approaches was performed. A 7-F long sheath was inserted into the right femoral artery, a 5-F long sheath was inserted into the left femoral artery, and a 6-F long sheath was inserted into the right femoral vein. Systemic heparinization was conducted in order to maintain the activated coagulation time at >250 during surgery. A 7-F Roadmaster (Goodman, Aichi, Japan) was guided into the right ICA and a 4F OK-1 (Gadelius Medical, Tokyo, Japan) was guided into the left ICA. A SHOURYU 7 mm × 7 mm (Kaneka Medix, Osaka, Japan) was guided to the petrous portion of the right ICA, and angiography of the left ICA and 3D rotational angiography were performed under balloon dilation. An Excelsior XT-17 (Stryker, Kalamazoo, MI, USA) was guided to a shunt segment, continuing from the aneurysmal neck, identified on the above procedures. The SHOURYU 7 mm × 7 mm was placed in the aneurysmal neck. Subsequently, a 6-F Roadmaster was guided into the right external jugular vein for transvenous embolization and a 4-F Cerulean G (Medikit, Tokyo, Japan) was guided into the right facial vein (**Fig. 3A** and **3B**). The venous drainage to the contralateral SOV and the facial vein was maintained even after guiding the 4-F catheter into the right facial vein (**Fig. 3C**). Based on the shape of the shunt segment, short-segment shunt occlusion was considered to be possible. However, if it was difficult, we planned to switch the procedure to sinus packing through transvenous embolization (TVE) after occluding the SMCV orifice. At this point, the facial vein approach was selected to occlude the reflux to the SOV, which caused the ocular symptoms. We considered stent-assisted coil embolization or ICA sacrifice at the second session if the shunted flow did not disappear despite the aneurysmal embolization. Regarding the Excelsior XT-17, which was guided to the shunt segment, as a landmark, an Excelsior SL10 (Stryker) was guided into the aneurysm through the 4-F Cerulean G, which was guided into the facial vein (**Fig. 3B**). Coils were inserted through the Excelsior XT-17 (guided through the ICA) and SL10 (guided through the SOV). A frame was formed using two coils (Penumbra SMART

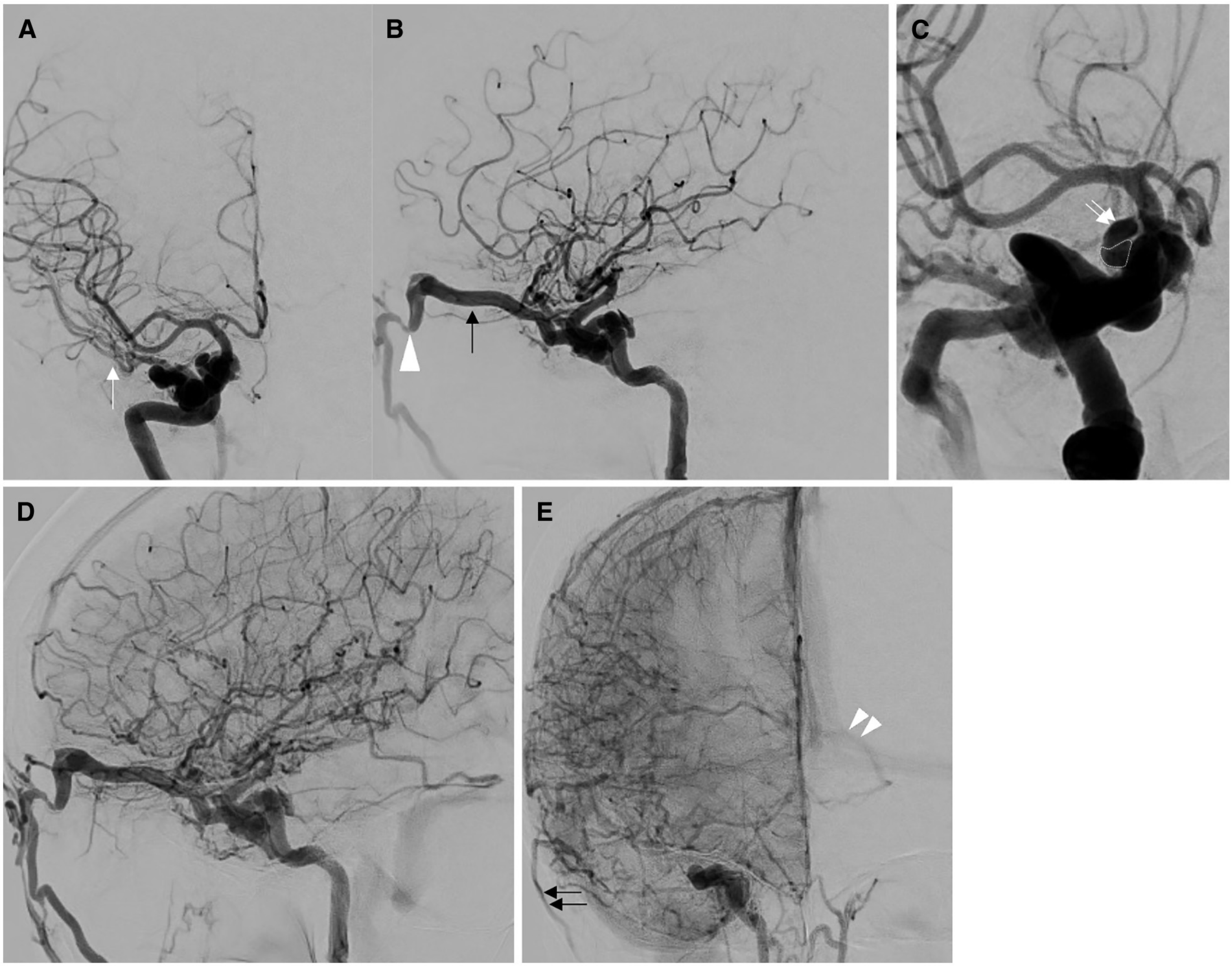

Fig. 2 DSA at diagnosis. (**A** and **B**) Arterial phase of initial right ICA angiography, anteroposterior (**A**) and lateral (**B**) views, showed a high-flow shunt at the cavernous sinus draining into the SOV (black arrow) and SMCV (white arrow). Furthermore, severe stenosis at the curve of an angular vein draining to the facial vein was revealed (white arrowhead). (**C**) DSA at the working angle to visualize the CCA clearly. The dotted line traces the contour of the aneurysm. White arrows indicate the shunt segment. (**D**) Late arterial phase on right ICA angiography showing cortical venous reflux with a pseudophlebitic pattern. (**E**) Early venous phase on right ICA angiography revealed venous drainage to the contralateral basal vein of Rosenthal (white arrowheads) and middle temporal vein (black arrows). CCA: carotid-cavernous aneurysm; ICA: internal carotid artery; SMCV: superficial middle cerebral vein; SOV: superior ophthalmic vein

SOFT 4 mm × 10 cm [Penumbra, Alameda, CA, USA] and ED complex 4 mm × 12 cm [Kaneka Medix]) by combining the double catheter technique with the neck remodeling technique by SHOURYU inflated. At first, the Penumbra SMART SOFT, which was guided through the transarterial catheter, was detached. For tight packing of the upper stream of the shunt, a Galaxy G3 XSFT 3.5 mm × 5 cm (Johnson & Johnson, New Brunswick, NJ, USA) and Hydrocoil 3D 2 mm × 3 cm (Terumo, Tokyo, Japan) were inserted under balloon inflation through the transarterial catheter. Angiography through the right ICA confirmed a marked decrease in shunted flow. However, to extirpate the shunt, the Galaxy G3 MINI 1.5 mm × 3 cm and 1.5 mm × 2 cm coils (Johnson & Johnson) were inserted through the transvenous catheter. A total of six coils were inserted through the two catheters guided via the transarterial and transvenous routes, and the shunt segment corresponding to the aneurysmal neck was tightly embolized (**Fig. 3D**). Final angiography showed complete obliteration of the direct CCF (**Fig. 3E** and **3F**).

Postoperative course: After surgery, conjunctival chemosis disappeared. The right ocular pressure was 9 mmHg, being lower than that before surgery (23 mmHg). Exophthalmos reduced from 17 mm before surgery to 13 mm after surgery. A modified Rankin scale at the discharge was 0. Follow-up by MRI and MRA of the head was continued. There has been no recurrence during the 2-year postoperative follow-up (**Fig. 3G**).

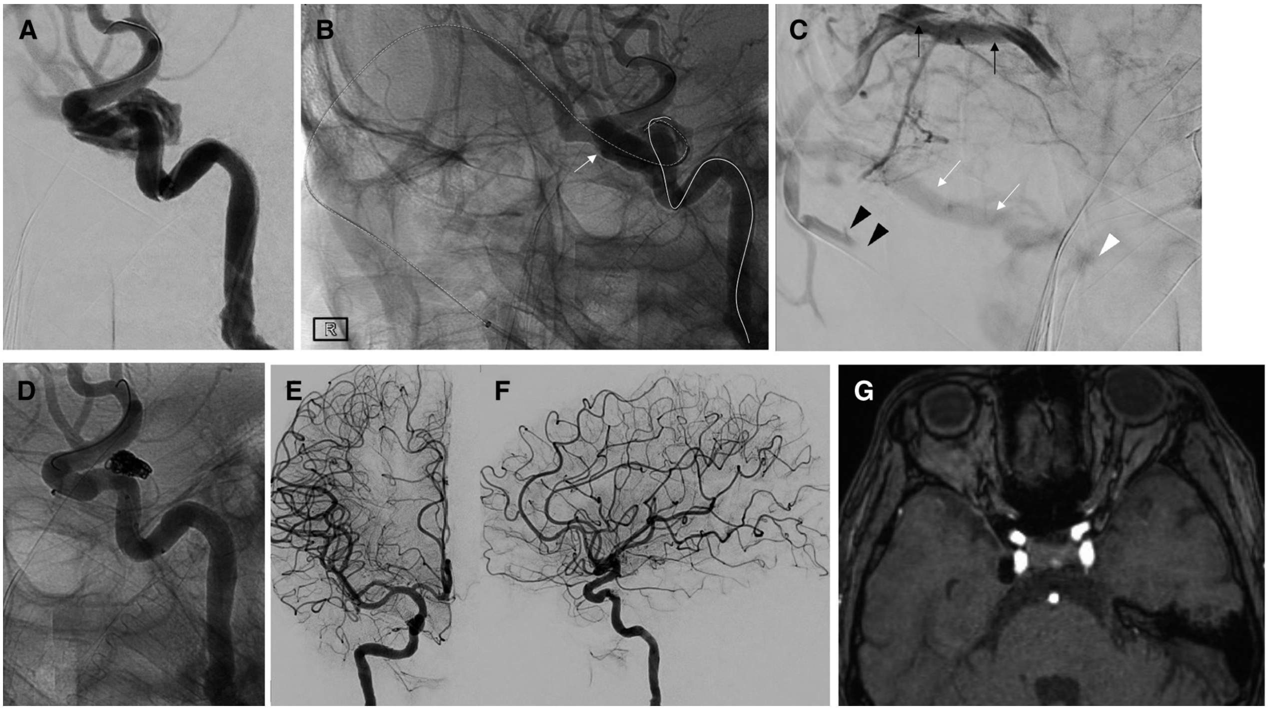

Fig. 3 Intraoperative angiography. (**A** and **B**) Working angle on right ICA angiography during the procedure, viewed from the left cranial projection (LAO 90 degree, Cranial 27 degree). The two microcatheters navigated to the fistula and intracranial balloon catheter (arrow) are shown in (**B**). Solid line: XT17 from the ICA. Dotted line: SL10 from the SOV. (**C**) Venous phase at the same working angle. Right ICAG after navigating the distal access catheter and microcatheter through the right SOV (black arrows) revealed sufficient venous drainage via the contralateral SOV (white arrows) to the cavernous sinus (white arrowhead) instead of the stagnant angular vein (black arrowheads). (**D**) The shunt segment is completely embolized with seven coils. (**E**) Right IC angiography, anteroposterior (**E**) and lateral (**F**) views, showed extirpation of the shunted flow. (**G**) Follow-up MRA 2 years after the procedure showed no recurrence of the aneurysm or the shunt. IC: internal carotid; ICA: internal carotid artery; ICAG: internal carotid angiography; LAO: left anterior oblique; SOV: superior ophthalmic vein

Discussion

CCAs account for 2 to 9% of intracranial aneurysms. Most CCAs are asymptomatic and incidentally diagnosed.[2,4] The incidence of rupture is lower than with other intracranial aneurysms. It is 0% for aneurysms measuring ≤12 mm and ranges from 3.0 to 6.4% for large aneurysms.[5] Although cranial nerve palsy is a popular symptom, direct CCF or subarachnoid hemorrhage owing to aneurysmal rupture occurs in some cases.[2,3] Ocular symptoms such as exophthalmos or conjunctival chemosis related to orbital congestion are primarily observed in direct CCF.[2,3] In the present case, direct CCF was diagnosed based on conjunctival chemosis and exophthalmos, which persisted for 3 months. However, the patient had a 1-year history of tinnitus, suggesting a long duration of disease. There was an increase in the right ocular pressure, and cerebral angiography demonstrated a pseudophlebitic pattern, which is a risk factor for intracranial hemorrhage.[6] Therefore, early shunt obliteration was considered to be necessary to prevent the loss of vision and intracranial hemorrhage.

Direct surgery for direct CCF, including parent artery occlusion, is highly invasive and the incidence of complications is high. For this reason, endovascular treatment has been selected as a first-choice treatment for direct CCF since Serbinenko reported treatment with a detachable balloon in 1974.[1,7,8] However, detachable balloons have technical problems, and the risk of recurrence is noted. In addition, these balloons are not available in several countries, including Japan[1]; therefore, endovascular treatment options for direct CCF include 1) aneurysmal occlusion, 2) cavernous sinus packing, and 3) parent artery occlusion. Intra-aneurysmal embolization of CCAs is recognized as the standard treatment.[3,8] The usefulness of stent-assisted coil embolization was also reported.[9] However, direct CCFs are often detected as high-flow shunts, finally requiring parent artery occlusion in approximately 15 to 20% of patients.[3,9] Recently, several case reports involving the use of a flow diverter were published, but the occlusion rate was not high.[10]

Aneurysmal occlusion or sinus packing may induce transient or permanent cranial nerve palsy related to a coil mass. The coil volume is a risk factor for this complication.[11,12] Superselective shunt occlusion preventing cranial nerve palsy, which is embolization with a small volume of coils in a shunt segment or shunted pouch located just downstream of a shunt point for cavernous sinus dural arteriovenous fistula, was reported. Its results are favorable.[13] In the present case, a shunt segment was identified at a junction of aneurysmal neck and cavernous sinus. Therefore, we presumed that embolization targeting this segment was able to obtain shunt occlusion.

The double catheter technique, which was first reported by Baxter et al., enables stabilization of a coil mass.[14] Currently, it is one of common adjunctive techniques used for the embolization of cerebral aneurysms. In addition, we reported the multipronged catheter approach involving a combination of transarterial and transvenous approaches for direct CCF, vertebro-vertebral arteriovenous fistula, and persistent primitive trigeminal artery-cavernous sinus fistula.[15–17] This method is useful for accurately guiding a microcatheter to a shunt segment, accomplishing compact, tight coil embolization, and arranging a risk hedge for unexpected catheter deviation. In the present case, bidirectionallyguided microcatheters through the ICA and SOV enabled embolization of the shunt segment with a minimum number of coils in spite of a high-flow shunt.

The inferior petrosal sinus should be selected as a first-line approach route to the cavernous sinus, because it is the shortest route even when it is occluded as reported for indirect CCF. SOV is important venous drainage in CCF, and the trans-SOV approach has been reported as one of the options for approaching cavernous sinus. However, the SOV approach may disturb the venous drainage during treatment. A previous study described the risk of exacerbating ocular symptoms or inducing intracranial hemorrhage due to the increased venous pressure in the cavernous sinus or other drainage route triggered by the SOV approach.[18] An SOV approach should be carefully determined. In the present case, the SOV approach was selected as it was anatomically advantageous from the viewpoint of microcatheter guiding to the shunt segment. A distal access catheter was guided proximal to the stenotic site of the angular vein avoiding obstruction of drainage route. Moreover, angiography during treatment revealed venous drainage to the contralateral SOV and no sign of increased venous pressure in the cavernous sinus or cortical vein. These findings were important for confirming the safety of the SOV approach.[18]

Conclusion

In a patient with a direct CCF related to CCA rupture, complete obliteration of the shunt was achieved by the double catheter technique from two directions: the ICA and the SOV. This bidirectional double catheter technique was safe and useful for embolization of the shunt segment with a minimum number of coils.

Disclosure Statement

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The other authors declare no conflicts of interest.

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